Traumatic cerebrospinal fluid fistulas in children

Departments of Radiology and Neurology, Adelaide Children's Hospital, North Adelaide, Australia

The postnatal development of the skull base was studied by dissection and radiography in 15 subjects, and the findings were correlated with the occurrence of traumatic cranionasal and crainioaural cerebrospinal fluid fistulas in seven children. The authors conclude that indications for operative repair of these fistulas are as strong in the child as in the adult, although diagnosis is sometimes more difficult.

KEY WORDS - cerebrospinal fluid - rhinorrhea - otorrhea - skull base - children - head injury

Cerebrospinal fluid (CSF) rhinorrhea and otorrhea are common sequelae of fractures of the skull base. They are clinically important as evidence of traumatic fistulas into the nasal or tympanic cavities, and are often complicated by meningitis or cerebral abscess. Fistulas into the nasal cavity heal so badly, and are so often the cause of early or delayed CSF infections, that many surgeons regard rhinorrhea, however transient, as an indication for exploratory craniotomy and dural repair. Others are more conservative, and advise operation only when the fluid leakage persists for 2 weeks, or where there is a history of prolonged or recurrent meningitis, or on the basis of detailed radiographic examination. Fistulas into the middle ear apparently do better and are more commonly treated expectantly, although delayed meningitis is not unknown.

There is still controversy over the management of these fistulas. Of the writers whose views have been influential, the majority have dealt chiefly with adult cases, although some have indicated that their principles of management are valid at all ages. Writers of text books concerned specifically with head injury in children have given lucid accounts of the occurrence of CSF fluid leakages, but their recommendations on management are not always in agreement. Mealey and Matson felt that rhinorrhea usually ceases spontaneously within a week, and advocated a conservative approach, with prolonged broad spectrum antibiotic cover for "several weeks." Moyson and Podevain more recently suggested a delay of 15 to 21 days before considering neurosurgical intervention. Dugger felt that even when rhinorrhea ceases spontaneously there is a high risk of subsequent infection; he recommended bifrontal craniotomy and dural repair as soon as the general condition of the child would permit. He considered, however, that otorrhea "almost never" required surgical closure.

Grote, in one of the few published articles dealing specifically with this problem in early life, described 14 cases of CSF fistulas...
in children ranging in age from 7 to 16 years at the time of operation. He gave numerous indications for surgical intervention, including both prolonged and transient rhinorrhea and aerocele, and even advised surgery where these were lacking but a significant bone defect could be seen in radiographs. He stressed, however, the diagnostic difficulties that may arise with children.

CSF fistulas are not very rare. Hendrick, et al., recorded 39 cases (rhinorrhea in 17 and otorrhea in 22) in a series of 4465 consecutive head injuries in children aged 14 or less.

It is the purpose of this paper to consider first the anatomy of the developing skull in relation to traumatic cranionasal and cranioaural fistulas and, second, to describe selected clinical experiences that seem to justify the policy of operative management advocated by Lewin, Grote, and others, even in the very young.

Anatomical Study

Purpose of Study

For the present purpose, there are two aspects of the anatomy of the developing skull base to be considered. The first is its structural composition: is the immature cranial base likely to fracture in the same way as does the adult skull? The second concerns the postnatal development of the middle ear cleft, the nose, and the paranasal sinuses: since some of these cavities do not reach their maximum development until adult life, is the risk of a traumatic fistula lessened in the infant or in the young child? These practical considerations have received relatively little attention. Gurdjian and Webster do note that the infant's skull, being relatively pliable in its vault although rigid in its base, is less likely to suffer linear fractures than the adult skull; after 2 or 3 years, the vault becomes rigid, and fracture lines conform more closely to the expected adult pattern. These writers briefly mention the development of the air sinuses, but do not correlate this with the liability to CSF fistulas.

Method

We have performed a number of postmortem dissections to amplify the standard anatomical and radiographic accounts of the development of the skull base. In all, 15 subjects were dissected, the ages ranging from birth to 75 years. In 12 of these dissections, a uniform procedure was used: the dura was stripped from the anterior cranial fossa, and the cribriform plates were removed. The air sinuses (except the maxillary antra) were then exposed from above, to the extent that they were in close relation (5 mm distant) to the anterior cranial fossa.

The resulting defect was photographed and also delineated in radiographs by a viscous medium. The state of the sutures and the presence of cartilage were noted.

In three other subjects, the procedure was varied to permit removal of the ethmoid region en bloc, for decalcification and serial histological section in the coronal plane. In one subject, the mastoid air cells and tympanic cavity were serially sectioned in the sagittal plane. The histological sections were stained with hematoxylin and eosin, and measurements of the thickness of the tissues between the nasal or paranasal cavities and the inner surface of the dura mater were made with an eyepiece graticule.

The following account is based on observations made in these dissections, supplemented by standard texts and other published reports, notably the excellent accounts given by Shapiro and Janzen and Frazer.

Probst's important work on frontobasal head injuries had not been published when our studies were carried out. This monograph gives an excellent summary of the relevant anatomical data, the statistics on age variations being particularly helpful.

Developmental Anatomy of the Anterior Cranial Fossa

The anterior cranial fossa comprises the frontal bones, which develop by ossification in membrane: the ethmoid bone, which is formed by ossification in the cartilages of the nasal capsules and nasal septum; and the very complex sphenoid bone, which has elements formed in membrane, but which is largely ossified in cartilage.

Frontal Bones. At birth, the frontal bones are well formed: thin plates of bone cover the orbits and articulate with each other and with adjacent bones by membranous sutures. These sutures are at first quite wide; the interfrontal (metopic), for example, measures...
Traumatic cerebrospinal fluid fistulas in children

1 to 2 mm at birth. They progressively narrow and interdigitate. The metopic suture usually disappears entirely during the second year of life; the other sutures remain open until adult life, but they have become so interlocked by the fourth year that fracture lines commonly run across them (Fig. 1). The frontal air sinuses arise as extensions either from anterior ethmoidal air cells or directly from the middle nasal meatus. We have not been able to identify these sinuses in young infants; it is usually stated that they become evident during the third year, invading the frontal bones progressively to develop in adult life a more or less extensive relationship to the anterior cranial fossa.\textsuperscript{22} In many cases, however, growth is slower, and the frontal sinuses do not constitute a surgical relationship to the anterior fossa until the age of 4 or even older.\textsuperscript{19} The risks of CSF fistulas into these sinuses do appear to be negligible until older childhood, at least in the majority of cases (Fig. 2).

**Ethmoid Bones.** The ethmoid bone is partly cartilaginous at birth. Its lateral components are the nasal capsules which develop into the facial component of the ethmoid bone, and are fully ossified at birth.\textsuperscript{22} However, the central component, the perpendicular plate, which includes the crista galli, the medial portions of the cribriform plate, and part of the septum, is almost wholly cartilag-

![Fig. 1. Postmortem photograph showing a skull-base fracture in a 3½-year-old boy. The fracture line (arrows) runs through the frontal, sphenoid, and petrous temporal bones; neither the membranous sutures nor the basilar synchondrosis show diastasis. The nasal cavities, ethmoid air cells (m), and small frontal air cells have been opened.](image)

![Fig. 2. Diagrammatic demonstration of the surgical relationships of the pneumatized cavities in the skull base. Drawn from postmortem photographs. Left: 5 months. Center: 5 years. Right: adult.](image)
inous at birth (Fig. 3). Ossification begins in the septum soon after birth, however, and during the first year the cribriform plate progressively ossifies; some cartilaginous areas persist as late as 2 years, but by this time the cribriform plate is almost wholly composed of bone. Thus, at birth the ethmoid component of the anterior fossa is composed of thin cartilage, which is more flexible than bone; over the next 2 years this is transformed into still thinner and more rigid bone, which presumably is apt to shatter. In specimens studied by us, the cartilaginous cribiform plate was about 800 μ thick at birth; the bony cribiform plate was much thinner at 3 years, being in some areas less than 100 μ thick.

The nasal cavity is, of course, in direct relationship to the anterior fossa at birth, in two linear strips about 20 to 25 mm long (Fig. 2). The nasal fossae are separated from the dura of the anterior fossa only by mucosa and cartilage, through which pass the filaments of the olfactory nerves (Fig. 3). This area of contact grows very little in later life, being only 25 to 30 mm long in older children; it is very narrow at birth, but broadens a little later.

The ethmoid air cells are clearly evident at birth as one or two small cavities in the ossified lateral components of the ethmoid bone. While they are more capacious than would be supposed from the description ("dimples") given by Hamilton, et al., they certainly do not bear any surgical relationship to the anterior fossa. They expand rapidly, however, and in a 5-month-old infant they are in direct relationship to the dura, being separated by a thin plate of bone (Fig. 2). By the third year, these cells are quite capacious, and greatly broaden the area through which fistulas could develop to a rectangle about 25–30 × 15–20 mm; by 10 years of age, this area is a little broader but of about the same length.

Thus, there is a theoretical risk of traumatic CSF fistula into the nasal cavity even at birth, but it is probably reduced by the cartilaginous nature of the cribiform plate. As this structure ossifies, and as the ethmoid air cells expand, the risk becomes more definite. By the third year, the nasoethmoidal cavities are relatively as large as those in the adult, and anatomical considerations suggest that skull base fractures might easily lead to cranionasal fistulas as, in fact, they do.

Sphenoid Bone. The sphenoid bone develops by endochondral ossification in two main components, the presphenoid and the postsphenoid. At birth, these units are usually fused, being joined posteriorly to the basisphenoid by a synchondrosis, and anteriorly to the ethmoid cartilage by a rather broad fibrous suture; the bone is thus largely ossified, with, however, more flexibility than in later life.

The sphenoid air sinus is said by Hamilton, et al., to be present at birth. If so, it is very diminutive; it certainly expands slowly, and is the last of the air sinuses to develop a significant surgical relationship with the anterior cranial fossa. By 5 years, it is usually well seen; however, it is commonly separated from the dura by cancellous bone so thick that it would preclude fistula formation. By 10 years, however, the sinus is often capacious, and is in relation to the dura in a broad area, perhaps 30 mm across. This sinus shows considerable individual variation.

Developmental Anatomy of the Middle Cranial Fossa

The middle fossa is formed by the sphenoid bone, which in this area ossifies partly in cartilage and partly in membrane, and the temporal bone, whose petrous component ossifies in cartilage.

Sphenoid Bone. The development of the sphenoid has been briefly discussed: it articulates with the petrous bone by a fibrous suture, and with the basisphenoid by the synchondrosis. This part of the skull base is more rigid than the anterior fossa. The sphenoid air sinus may develop a relationship with the middle fossa dura in older children and adults, if it is large enough to extend lateral to the cavernous sinus; we have, however, not noted this relationship in our pediatric material.

Petrous Bone. The petrous bone develops from several centers in the cartilaginous otic capsule, but by birth it is wholly ossified. The middle ear cleft is large at birth; the tympanic cavity and the mastoid antrum have a substantial dural relationship over an area about 10 × 20 mm. The bone of the tegmen tympani is quite rigid, and very thin; the possibility of a CSF fistula into the
Traumatic cerebrospinal fluid fistulas in children

Fig. 3. Coronal histological sections, approximately ×4.5, showing the cribriform plate at birth (upper), at 3 months (center) and at 3 years (lower). The nasal cavities (N) are separated from the anterior cranial fossa and olfactory bulbs (OB) by tissues that are slightly thicker in the neonate than in older infants. The crista galli is composed of cartilage, staining darkly (alcein blue) in the neonate; at this age, the cribriform plate is also largely cartilaginous. By the third year, these structures are entirely ossified. Ethmoid air cells (E) are not in proximity to the anterior cranial fossa at birth, but have a limited relationship at 3 months and an extensive relationship at 3 years.
middle ear would therefore seem, on anatomical grounds, quite real, even in young infants. However, the mastoid air cells are diminutive at birth, and extend only gradually into widespread relationship with the dura. By 5 years, the relationship is often quite extensive; moreover, the mastoid air cells may establish a dural relationship in the posterior fossa, medial and lateral to the internal auditory meatus.

Case Reports

During the period 1953–1970, seven children were found at operation or autopsy to have definite traumatic fistulas in the skull base, resulting in CSF leakage, or meningitis, or a combination of these complications. Two other children were strongly suspected of having such fistulas, though they were not verified at exploratory craniotomy; Case 2 is one of these.

Cranioaural Fistula

Case 1. A girl aged 2 yrs 8 mos was injured in a road accident. She exhibited an abducens palsy and a CSF otorrhea, both on the right side. These disappeared spontaneously, and she remained well for 13 years. She then developed two severe attacks of pneumococcal meningitis which were treated with penicillin. The middle fossa was explored, and a small cerebral hernia was found, entering a defect in the tegmen tympani. The defect was repaired with fascia lata and a muscle stamp. She has remained well since.

Case 2. This case is not given in detail, as the operative findings were equivocal. An 11-year-old boy presented with profuse CSF otorrhea. Skull radiographs showed multiple fractures, one of which appeared to involve the internal auditory meatus, and also a small intracranial aerocele. The fluid leakage ceased spontaneously after two weeks.

Two months later, the patient developed acute meningitis, probably pneumococcal. Exploration of both middle and posterior cranial fossae showed no obvious fistula; however, the internal auditory meatus was packed with muscle stamps, in the presumption that it was the route of infection. The boy remains well, five years later.

Cranionasal Fistulas

Case 3. A 2 year-old boy was kicked by a horse in the right frontal region. He sustained a small compound fracture, which was repaired; he was reported to have leaked CSF from the nose, but this had ceased spontaneously. Tomograms of the anterior fossa showed fractures in both orbital roofs, involving the ethmoid air sinuses on the right side. Nevertheless, he was treated conservatively, both because of his youth and because the leakage of CSF was not verified.

Over the next 16 months, the child repeatedly suffered right-sided rhinorrhea. It was at first regarded as due to recurrent colds; eventually, however, test with Clinistix glucose indicator suggested that there was indeed a CSF leakage. Intrathecal injection of 131I human serum albumen gave an equivocal result. Bifrontal craniotomy showed extensive dural tears, on both sides; the right cribriform plate was apparently intact, but the left was disrupted by a jagged fracture. Both cribriform plates were covered with muscle and a large sheet of pericranium was sutured over them. He has been well over the subsequent year.

Case 4. A 9-year-old boy who had sustained a frontal head injury appeared to recover well, but 2 weeks later developed a definite right-sided CSF rhinorrhea. Radiographs showed a linear fracture running into a diminutive right frontal sinus, and probably also into the ethmoid sinuses. He had no anosmia. At operation, a fistula was seen; it was not clear whether it entered the frontal sinus or an anterior ethmoidal air cell. Temporalis fascia was sutured over the fistula. He remains well, 2 years later.

Case 5. A 9-year-old girl was struck on the forehead by a tree branch, and after treatment in a remote mission hospital appeared to recover. However, during the next 18 months she suffered two bouts of meningococcal meningitis. Bilateral orbital fractures were seen in radiographs, and at operation two fistulas were found, running into the ethmoidal air cells on the right. Fascia lata was sutured over the fistulas. She made a good recovery, and is now reported to be well, except for epileptic seizures.

Case 6. An 11-year-old girl sustained a frontal head injury in a bicycle accident. At first she seemed well, but on the following day developed fulminating meningitis, and despite antibiotic treatment, died. No CSF leak was recorded, but she was not under
our observation at first. Autopsy showed a fracture in the left orbital roof, running into the frontal and ethmoid sinuses and into the nasal cavity. The nature of the infecting organism was not established.

**Case 7.** An 11-year-old girl sustained a frontal head injury in a vehicular accident, she evidently hit the windscreen, and there was an extensive frontal skin wound running to the glabella. On admission, the house surgeon reported leakage of blood and CSF from the left ear; this was not noted later. Skull radiographs showed a linear frontal fracture running into the nasal region, without depression. The skin wound was sutured, and she made a good recovery. Five months later, she developed CSF rhinorrhea and this was followed by two severe attacks of meningitis; in the first, no organism was recovered, but in the second the organism was considered (in another hospital) to be staphylococcal. A general surgeon attempted a repair of the presumed frontal fistula, but the leakage persisted, and the girl suffered two more bouts of meningitis.

Nearly 2 years after her injury, a left frontal craniotomy was performed, exposing an extensive dural defect and a bone defect in the posterior wall of the left frontal sinus. This was covered with temporalis fascia. After this operation, the girl leaked CSF from the right nostril. A right frontal craniotomy was then performed, and this disclosed a second bone defect into the ethmoid air cells, which had not been seen in the skull radiographs. (Tomography was not at that time available in Adelaide.) This defect was covered with fascia lata.

The girl then seemed well, but 6 months later had a fifth attack of meningitis, this time certainly pneumococcal. A fourth craniotomy was performed, and this disclosed a second bone defect into the ethmoid air cells, which had not been seen in the skull radiographs. (Tomography was not at that time available in Adelaide.) This defect was covered with fascia lata.

The girl then seemed well, but 6 months later had a fifth attack of meningitis, this time certainly pneumococcal. A fourth craniotomy was performed, and this disclosed a second bone defect into the ethmoid air cells, which had not been seen in the skull radiographs. (Tomography was not at that time available in Adelaide.) This defect was covered with fascia lata.

**Case 8.** A 6-year-old boy was struck by an automobile and sustained a right frontal head injury. There was a deep skin laceration and under it a linear fracture with depression of some fragments of bone. This fracture ran into the right orbit and was thought to enter the right frontal air sinus. There were paralyses of the right second and third nerves. The wound was sutured by a general surgeon after elevation of the depressed fragments, and the child made a good recovery but 16 months later had a severe attack of pneumococcal meningitis. The frontal sinus was then explored by an aural surgeon, and a CSF leak into the posterior wall of this sinus seen. Nothing was done, and during the next 3 years the boy had two further attacks of meningitis. After the last, a neurosurgeon was consulted; he performed a right frontal craniotomy and found several small fistulas into the cribriform plate. These were covered with a fascial graft, and the boy made a good recovery. He was well 12 years later, apart from being blind in the right eye.

**Discussion**

Our experience confirms what many others have reported: that traumatic CSF fistulas are certainly not rare in childhood. In a period of 15 years, our neurosurgical group treated more than 70 fistulas. Our adult-child ratio of about 10 to 1 is much like that reported by others. Children under the age of 15 constitute about 30% of the population of our community.

We have not seen the case of a patient younger than 2 years of age, nor have we found any in the literature, with the exception of Grote's fifth patient who appears to have been 1 year old when injured. This apparent immunity of infants may represent the rarity of head injuries at this age, and particularly of direct impacts to the frontal region. But the flexibility of the skull base in infancy, and especially the cartilaginous nature of the ethmoid, may also be relevant.

In patients older than 2 years, fistulas have been seen both into the nasoethmoidal cavities and the middle ear. By 5 years the frontal air sinuses become progressively more significant as alternative sites of fistulas: in our Cases 4, 7, and 8, these sinuses were involved, and Grote reported frontal fistulas in four cases. We saw no sphenoidal fistulas in this series, with the possible exception of one of the fistulas found in Case 7. It
appears from the literature to be rare; the anatomical reasons for this have been given.

Recognition of CSF rhinorrhea and otorrhea may be difficult in children, especially if the leakage is transient. Runny noses are very common in this age group, and the child or the parent may not mention the complaint. A positive reaction for glucose by a test paper has been useful in our experience, but is not wholly unequivocal.\(^2,8\)

A radioactive tracer substance injected into the CSF by lumbar puncture will give elegant confirmation of the nature and even the site of a fistula\(^4\) if there is a continuing leakage; of course it will not do so if the leakage has temporarily ceased, as was the case in one of our children (Case 3).

As has been observed, the indications for surgical closure of a fistula are debatable. It was initially our opinion that a more conservative policy might be justified in children, on the assumption that basal fractures would heal more readily in the immature skull. This does not appear to be the case, at least in regard to anterior fossa fistulas. Our Case 3 shows that dangerous fistulas may persist even in the very young.

That such fistulas are dangerous, especially in age groups whose immunological defenses may be inadequate, is shown by our Case 5, where an early fulminating and fatal meningitis occurred before the presence of a fistula was suspected. Prophylactic chemotherapy may be a protection;\(^3\) but we consider that the risk of meningitis is substantial, not only in the early days, but also, as in our Cases 1, 5, 7 and 8, after long intervals. Meningitis is still a very dangerous disease; we have recently seen it cause death and crippling dementia in two adults whose fistulas might have been repaired earlier. It is therefore our policy to recommend exploration of the anterior fossa in all cases of CSF rhinorrhea (or rhinogenic aerocele), provided that the diagnosis is unequivocal. A conservative policy may be justified when the leakage is dubious or very transient, and when skull radiographs show no fracture. However, if a fracture is visible, then even a brief episode of rhinorrhea is an indication for exploration.

The indications for operation in CSF otorrhea are less compelling since there is no doubt that many cases do well without surgical treatment. But Cases 1 and 2 show that this is not always so. Previously, we have been reluctant to embark on operation unless compelled to, because of uncertainty as to whether to explore the middle or posterior fossa. Improvement in the technique of tomographic examination of the petrous bone\(^12\) should reduce this uncertainty. At present we advise exploration when unequivocal CSF otorrhea persists for 7 days; and if radiography has shown a large petrous bone defect, then even transient otorrhea (or escape of cerebral tissue) should be taken to justify surgery.

Radiographic demonstration of intracranial air has basically the same significance as CSF leakage, but it may at times be hard to establish the anatomical site of the causative fistula. Only one of our cases showed this radiographic sign.

Our policies would of course receive conclusive evaluation if we could report a control series of children treated expectantly. However, our earlier experiences of the risks of expectant treatment of CSF rhinorrhea were so convincing that we have not thought it proper to attempt such a comparison, though we have under long-term supervision two young children who sustained profuse CSF otorrhea, which ceased spontaneously; so far, both have been well.

Operation is, of course, deferred until the child has recovered from the cerebral insult. During the period of recovery, we have usually given penicillin and sulphadiazine in high dosage, unless culture of aural or nasal discharge has suggested the need for some other antibiotic. The choice of sulphadiazine has been criticized;\(^3\) this drug has the merit of good passage into the CSF, but perhaps an aminoglycoside such as gentamicin may be preferable.

Our technique of operative repair has been modelled on that of Lewin.\(^14\) Muscle stamps have been placed in the fistula, which is then covered by a wide sheet of fascia or pericranium. This is invariably sutured in position. Magnification is very helpful. The only recurrent fistula seen after this procedure (Case 7) occurred early in our experience; a wider bilateral exposure would now be the initial operation, and unilateral exploration is done only when the radiographic identification of the fistula seems conclusive.
Traumatic cerebrospinal fluid fistulas in children

Transaural or transnasal repair may be feasible, but it seems to us that these approaches plug the fistulas from the wrong side, and are therefore less likely to be reliable.

Acknowledgments

We thank Mr. T. A. R. Dinning, F.R.C.S., F.R.A.C.S., formerly honorary neurosurgeon to this hospital, for permission to report five of the cases in this paper, and for much advice in its preparation.

References


Address reprint requests to: Donald Simpson, M.S., F.R.C.S., F.R.A.C.S., Department of Neurology, Adelaide Children's Hospital, 72 King William Road, North Adelaide, 5006, Australia.